

Middle meningeal arteriovenous fistulas: A rare and potentially high-risk dural arteriovenous fistula

Rami O. Almefty, M. Yashar S. Kalani, Andrew F. Ducruet, R. Webster Crowley, Cameron G. McDougall, Felipe C. Albuquerque

Department of Neurosurgery, Barrow Neurological Institute, St. Joseph's Hospital and Medical Center, Phoenix, Arizona, USA

E-mail: Rami O. Almefty - neuropub@dignityhealth.org; M. Yashar S. Kalani - neuropub@dignityhealth.org; Andrew F. Ducruet - neuropub@dignityhealth.org; R. Webster Crowley - neuropub@dignityhealth.org; Cameron G. McDougall - neuropub@dignityhealth.org; *Felipe C. Albuquerque - neuropub@dignityhealth.org
*Corresponding author

Received: 16 September 15 Accepted: 29 January 16 Published: 01 April 16

Abstract

Background: Middle meningeal arteriovenous fistulas (MMAVFs) are rare lesions with a poorly established natural history. We report our experience with patients with MMAVFs who presented with intracranial hemorrhage.

Methods: We reviewed our prospectively maintained endovascular database for patients with MMAVFs, who were treated by embolization during a 15-year period. Hospital and outpatient medical records and imaging studies were reviewed.

Results: Nine patients with MMAVFs, who presented with intracranial hemorrhage, underwent embolization (mean age 60.3 years, range 21–76; four male and five female). Four patients presented after trauma and five after spontaneous hemorrhage. All nine patients were angiographically cured after embolization of the fistula with liquid embolic agents ($n = 8$) or coils ($n = 1$). There were no procedure-related complications.

Conclusion: MMAVFs represent a rarely reported class of vascular lesions. They are typically associated with trauma, but also develop spontaneously, and may be associated with intracranial hemorrhage, which warrants classification of these lesions as high risk. Endovascular treatment is safe and effective and should be considered for these patients, particularly for those who have lesions with intracranial venous drainage.

Key Words: Arteriovenous, embolization, fistula, hemorrhage, meningeal, vascular

Access this article online

Website:

www.surgicalneurologyint.com

DOI:

10.4103/2152-7806.179575

Quick Response Code:



INTRODUCTION

Middle meningeal arteriovenous fistulas (MMAVFs) occur between the middle meningeal artery and neighboring veins or dural venous sinuses and are rare lesions that have largely been the subject of single case reports.^[1-16] As many patients with MMAVFs remain asymptomatic, the true incidence of this type of lesion remains unclear, making the natural history of these lesions uncertain. Although MMAVFs are typically associated with trauma, spontaneous cases have also been reported.^[1,6] Our series indicates that regardless of the etiology of the fistulas,

these lesions pose a risk of intracranial hemorrhage and treatment should be considered. The middle meningeal

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Almefty RO, Kalani MY, Ducruet AF, Crowley RW, McDougall CG, Albuquerque FC. Middle meningeal arteriovenous fistulas: A rare and potentially high-risk dural arteriovenous fistula. *Surg Neurol Int* 2016;7:S219-22. <http://surgicalneurologyint.com/Middle-meningeal-arteriovenous-fistulas-A-rare-and-potentially-high-risk-dural-arteriovenous-fistula/>

artery is typically accessible through an endovascular approach, making these lesions highly amenable to endovascular cure.

METHODS

We searched our prospectively maintained endovascular treatment database for patients with MMAVFs, who were treated by embolization. Only those cases in which a fistula existed between the middle meningeal artery and neighboring veins or dural sinuses were included. Cases in which the middle meningeal artery contributed to dural arteriovenous fistulas of the major venous sinuses were not included. Hospital and outpatient medical records and imaging studies were reviewed for patients who met the inclusion criteria.

RESULTS

Nine patients (five female and four male) with MMAVFs [Table 1] underwent endovascular treatment at our institution between January 1, 2000, and April 30, 2015. The average age was 60.3 years (range, 21–76 years). Four patients exhibited evidence of traumatic injury, although in two cases the patients were found unconscious and the mechanisms and circumstances of injury remained uncertain. All patients presented with intracranial hemorrhage: Three with isolated subarachnoid hemorrhage (SAH), four with primarily intraparenchymal hemorrhage (IPH), and two with multicompartmental hemorrhage. The majority of patients presented with mental status change or a depressed level of consciousness. All fistulas were embolized without complications using N-butyl-cyanoacrylate (nBCA; six cases), Onyx (ev3/Covidien, Irvine, CA, USA; two cases), or coils (one case). Complete obliteration was achieved in all cases. Only four patients with IPH had focal deficits that corresponded to the site of hemorrhage. In each case, these deficits persisted following treatment. The remaining five patients were neurologically intact at discharge or short-term follow-up.

Case Illustration

A middle-aged patient was admitted to our service after being involved in a serious accident. On presentation to the trauma bay, the patient’s Glasgow Coma Scale score was 9. Given this poor score and respiratory distress, the patient was intubated. Imaging revealed traumatic SAH [Figure 1a] and a pneumothorax for which a chest tube was placed. Other injuries included fractures of the left tibia and fibula, multiple facial fractures, and a T7–T8 vertebral fracture.

As a part of routine work-up for cases of traumatic brain injury, computed tomography angiography of the head was ordered. Review of this study suggested the presence of a small aneurysm arising from the left internal carotid artery [Figure 1b]. The patient was stabilized and resuscitated. After discussion among the trauma, orthopedic, and plastic surgery services, it was decided that the patient should undergo formal angiography to evaluate the cerebral circulation and to rule out a vascular etiology contributing to the patient’s hemorrhage.

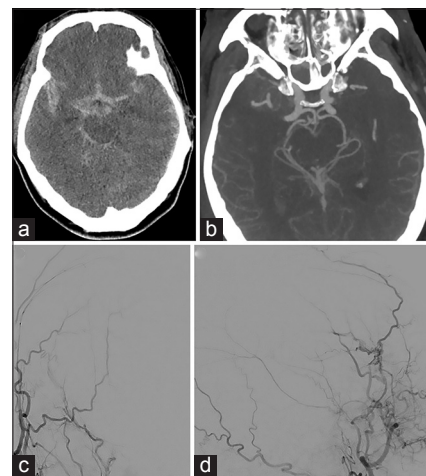


Figure 1: Initial diagnostic tests after trauma. (a) Axial computed tomographic scan shows thick, diffuse subarachnoid hemorrhage. (b) Computed tomographic angiogram shows left internal carotid artery aneurysm. Anteroposterior (c) and lateral (d) angiograms show a middle meningeal artery-to-vein fistula. Used with permission from Barrow Neurological Institute, Phoenix, AZ, USA

Table 1: Demographics, imaging findings, and outcomes for 9 cases of middle meningeal arteriovenous fistula

Age (yr)	Presentation	Hemorrhage type	Embolysate	Obliteration	Outcome (mRS)	Trauma*
50s	Found unconscious	SAH	nBCA	Complete	1	–
70s	Found unconscious (+), stigmata of trauma	SAH	nBCA	Complete	1	+
70s	Headache, nausea, and vomiting	SAH/IPH	nBCA	Complete	1	–
20s	Status post-assault	SDH	Coils	Complete	1	+
70s	Found unconscious (+), stigmata of trauma	SAH	nBCA	Complete	1	+
50s	Aphasia and hemiplegia	IPH	nBCA	Complete	4	–
60s	Hemiplegia and lethargy	IPH	Onyx	Complete	4	–
70s	Aphasia and hemiplegia	IPH	Onyx	Complete	3	–
60s	Altered mental status	IPH	nBCA	Complete	3	+

IPH, intraparenchymal hemorrhage, mRS, modified Rankin Scale Score, nBCA, N-butyl-cyanoacrylate, SAH, subarachnoid hemorrhage, SDH, subdural hematoma.

*Skull fracture associated with case (+, yes; –, no)

Formal cerebral angiography, including selective injections of the bilateral internal and external carotid arteries and the vertebrobasilar system, was performed. The patient was noted to indeed have a small 2-mm aneurysm of the left internal carotid artery arising from the cavernous segment (not shown). The patient was also noted to have a traumatic arteriovenous fistula of the middle meningeal artery [Figure 1c and d]. No treatment was planned, and the patient was returned to the Intensive Care Unit. The patient's airway was transitioned to a tracheostomy, and the patient was further resuscitated.

Ultimately, the patient's neurological status remained poor with the patient spontaneously moving the right upper extremity and flexing the bilateral lower extremities (confounded by the presence of bilateral orthopedic injuries), and was flaccid in his left upper extremity.

Sixteen days after the initial trauma, the patient was noted to be less responsive, and a new computed tomography study revealed a new IPH [Figure 2a]. Computed tomography angiography demonstrated vasospasm in the right middle cerebral artery distribution and abnormal dilatation of a vessel at the right temporal tip [Figure 2b]. The patient was taken back to the angiography suite for repeat studies. Selective injection of the right external carotid artery revealed two new arterial pseudoaneurysms adjacent to the site of the original MMAVF [Figure 2c and d]. Given the proximity of the pseudoaneurysms to the site of hemorrhage and their intradural location, we elected to embolize these lesions with nBCA. Selective embolization of these lesions was

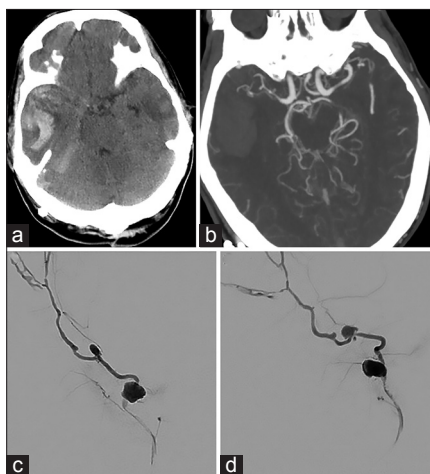


Figure 2: Imaging performed 16 days posttrauma after the patient experienced neurological decline. (a) Axial computed tomographic scan shows a new right temporal intraparenchymal hemorrhage. (b) Computed tomographic angiogram shows vasospasm in the right middle cerebral artery distribution and an abnormal dilatation in a vessel adjacent to the hematoma. (c and d) Selective middle meningeal artery injections show newly developed pseudoaneurysms associated with the previously detected fistula. Used with permission from Barrow Neurological Institute, Phoenix, AZ, USA

performed without complication [Figure 3]. The patient was discharged to a care facility with a stable neurological status with no additional follow-up.

DISCUSSION

Nine patients with MMAVFs and intracranial hemorrhage underwent endovascular fistula embolization. The cohort includes patients with fistulas of traumatic etiology and patients who presented with spontaneous intracranial hemorrhage and were found to have an MMAVF. Five of the 9 patients (63%) had no history of trauma – 1 patient presented with SAH, 3 had an IPH, and 1 presented with both SAH and IPH. It is well known that traumatic MMAVFs occur although the true incidence is unclear given that many patients may go undiagnosed. MMAVFs in patients presenting with spontaneous hemorrhage have only rarely been reported. Although the etiology of traumatic and spontaneous MMAVFs is different, we grouped them together because of their shared anatomical features, hemorrhagic potential, and treatment.

In the largest reported series to date, Freckmann *et al.* reviewed the angiograms of 446 head trauma patients and found eight cases of MMAVFs, for a rate of 1.8%.^[3] None of these patients were treated, and the series preceded the endovascular era. No other large case series have been reported, and the lesion has largely been the subject of single case reports.

Although the majority of MMAVFs occur after traumatic injury, some develop spontaneously. More importantly, as we observed in our patients without a prior history of trauma, these lesions pose a risk of intracranial hemorrhage, regardless of their etiology. As a result,

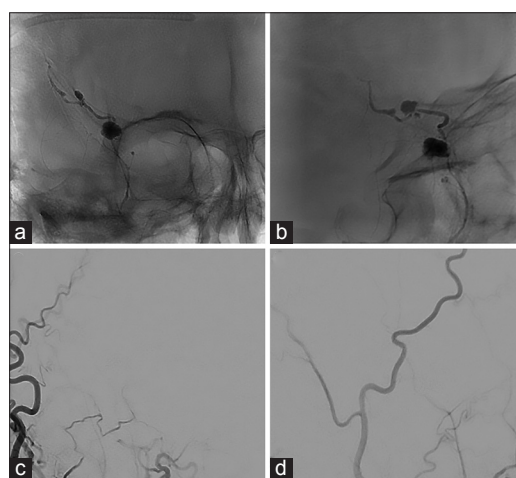


Figure 3: Imaging performed after embolization of the middle meningeal arteriovenous fistula with N-butyl-cyanoacrylate glue. (a and b) Unsubtracted views show glue cast in the fistula. Anteroposterior (c) and lateral (d) angiograms after embolization show no residual fistula. Used with permission from Barrow Neurological Institute, Phoenix, AZ, USA

the presence of an MMAVF may explain some cases of nonaneurysmal SAH and thus a complete work-up for SAH requires 6-vessel angiography.

Although MMAVFs have been rarely reported in the literature, the hemorrhagic potential of these lesions was clearly demonstrated by Sakata *et al.*^[10] In their report, a 48-year-old woman with a history of head trauma developed an MMAVF with dilated intracranial venous drainage that progressed on serial angiography. She later presented with diffuse, thick, basal SAH. The authors attributed this hemorrhage to the formation and rupture of a venous aneurysm associated with the draining vein. Our case illustration of a patient with MMAVF who developed an associated pseudoaneurysm found on serial angiography, which ruptured and caused a devastating IPH, further clarifies the potential dynamic and high-risk nature of these lesions.

Our experience certainly indicates that these lesions warrant consideration as high risk, particularly those with cortical venous drainage. We do not have a cohort of untreated MMAVF patients whom we have followed, which would give us a better sense of the natural history of these lesions. However, our series does demonstrate the hemorrhagic potential of MMAVFs as well as the ability to safely access and obliterate these lesions through an endovascular approach. On the basis of these findings, we believe that treatment of MMAVFs should be considered.^[1,4,16]

In our series, all fistulas were completely embolized without complications. Liquid embolic agents were primarily used given the small size and distal location of these fistulas. Although we have recently favored Onyx for transarterial embolization of dural arteriovenous fistulas, in cases where distal navigation of the microcatheter is limited by vessel tortuosity, we have found that a dilute mixture of nBCA may be more likely to effectively penetrate the fistula to achieve a definitive cure.

CONCLUSION

MMAVFs are rare and likely underreported vascular lesions. They are typically associated with trauma, but also develop spontaneously, and may lead to intracranial hemorrhage. They should be regarded as high-risk lesions and treatment should be considered, particularly for those lesions with intracranial venous drainage. Early treatment results for endovascular embolization are encouraging.

Acknowledgments

We thank the Neuroscience Publications Office at Barrow Neurological Institute for support.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Chandrashekar HS, Nagarajan K, Srikanth SG, Jayakumar PN, Vasudev MK, Pandey P. Middle meningeal arteriovenous fistula and its spontaneous closure. A case report and review of the literature. *Interv Neuroradiol* 2007;13:173-8.
2. Fincher EF. Arteriovenous fistula between the middle meningeal artery and the greater petrosal sinus; case report. *Ann Surg* 1951;133:886-8.
3. Freckmann N, Sartor K, Herrmann HD. Traumatic arteriovenous fistulae of the middle meningeal artery and neighbouring veins or dural sinuses. *Acta Neurochir (Wien)* 1981;55:273-81.
4. Ishii R, Ueki K, Ito J. Traumatic fistula between a lacerated middle meningeal artery and a diploic vein; case report. *J Neurosurg* 1976;44:241-4.
5. Liu Ah, Lv X, Li Y, Lv M, Wu Z. Traumatic middle meningeal artery and fistula formation with the cavernous sinus: Case report. *Surg Neurol* 2008;70:660-3.
6. Markham JW. Arteriovenous fistula of the middle meningeal artery and the greater petrosal sinus. *J Neurosurg* 1961;18:847-8.
7. Nakamura K, Tsugane R, Ito H, Obata H, Narita H. Traumatic arterio-venous fistula of the middle meningeal vessels. *J Neurosurg* 1966;25:424-9.
8. Pakarinen S. Arteriovenous fistula between the middle meningeal artery and the sphenoparietal sinus. A case report. *J Neurosurg* 1965;23:438-9.
9. Roski RA, Owen M, White RJ, Takaoka Y, Bellon EM. Middle meningeal artery trauma. *Surg Neurol* 1982;17:200-3.
10. Sakata H, Nishimura S, Mino M, Hori E, Fujita T, Midorikawa H, *et al.* Serial angiography of dynamic changes of traumatic middle meningeal arteriovenous fistula: Case report. *Neurol Med Chir (Tokyo)* 2009;49:462-4.
11. Sicut LC, Brinker RA, Abad RM, Rovit RL. Traumatic pseudoaneurysm and arteriovenous fistula involving the middle meningeal artery. *Surg Neurol* 1975;3:97-103.
12. Smith JE, Epps J, Press HC Jr., Adair LB. Traumatic arteriovenous fistula between the middle meningeal artery and the sphenoparietal sinus: A case report and review of the world literature. *J Natl Med Assoc* 1981;73:274-8.
13. Tsutsumi M, Kazekawa K, Tanaka A, Ueno Y, Nomoto Y, Nii K, *et al.* Traumatic middle meningeal artery pseudoaneurysm and subsequent fistula formation with the cavernous sinus: Case report. *Surg Neurol* 2002;58:325-8.
14. Unterhofer C, Chemelli A, Waldenberger P, Bauer R, Ortler M. Traumatic fistula between the middle meningeal artery and the sphenoparietal sinus. *Acta Neurochir (Wien)* 2009;151:1301-4.
15. Watanabe T, Matsumaru Y, Sonobe M, Asahi T, Onitsuka K, Sugita K, *et al.* Multiple dural arteriovenous fistulae involving the cavernous and sphenoparietal sinuses. *Neuroradiology* 2000;42:771-4.
16. Wilson CB, Cronin F. Traumatic arteriovenous fistulas involving middle meningeal vessels. *JAMA* 1964;188:953-7.